

SCIREA Journal of Clinical Medicine http://www.scirea.org/journal/CM ISSN: 2706-8870 November 7, 2019

Volume 4, Issue 6, December 2019

Treatment for Unicystic Ameloblastoma by Adopting the Technique of Decompression by Fenestration with the Second Molar Preserved

Wen-chao Li^{1*}, Ning Ruan¹, Yuan Tian¹, Wei Zu¹, Hai-jie Guan¹, Yang Lin¹, min-Qiang Zhao¹, jiang Xu²

¹Department of Oral Surgery, Affiliated Hospital of Chifeng College, Chifeng, 024000, China

²Department of Stomatology, The First Affiliated Hospital of Medical College of Shihezi University, Shihezi, 832000, China

*Corresponding author: Wen-chao Li, Master of Medicine.

Mailing address: Department of Oral Surgery, Affiliated Hospital of Chifeng College, Chifeng, 024000, China

Address: 42 Wangfu street, Chifeng songshan district of Inner Mongolia, Chifeng, 024000, China

Abstract

Background: Unicystic ameloblastoma (UAB) is a relatively rare type of Ameloblastoma (AB) and often occurs in adolescents. The technique of decompression by fenestration can help to preserve the appearance and function of the jaws.

Purpose: The aim of this study is to validate the efficacy of the decompression by fenestration technique in the treatment for UAB with the second molar preserved.

Methods: The decompression by fenestration technique was employed to six patients suffering from the UAB with the mandibular second molar involved. The mandibular second molar was preserved in the surgery. These patients were followed up for five years to monitor the prognosis.

Results: Eight months after the decompression by fenestration, the tumors' size decreased by 61.62%, the bone mineral density of the mandible inferior border increased, and the formation of the trabeculae was observed; six months after the tumors were removed, the jaws made a full recovery and the function of the mandibular second molar was preserved; during the follow-up period of one to five years, there was no tumor reoccurrence and the mandibular second molar functioned properly.

Conclusion: The disfigurement was relieved and the functions of jaw and tooth were protected by the application of the fenestration with the second molar preserved and the tumor removal surgery.

KeyWords: Unicystic Ameloblastoma; Fenestration; Decompression; Second Molar

Introduction

Ameloblastoma (AB) is a common odontogenic jaw tumor and it is highly proliferative, invasive, and recurrent [1, 2]. Unicystic ameloblastoma (UAB) is a rare type of AB and often occurs in adolescents. Although UAB is a borderline tumor, its local invasiveness and recurrence is lower than other types of AB [3]. In the UAB patients, the teeth are usually embedded in the tumor[2]. Clinical treatment of UAB usually removes the teeth and the tumor simultaneously to prevent recurrence [4]. This surgery method usually causes the change of the face and decrease of the masticatory function due to the removal of the teeth and the bone tissues. It raises the difficulties of follow-up oral plastic surgery or oral implantation. The patients would also bear a serious psychological burden due to the possible disfigurement and the weakening jaw functions.

Conservative treatments for UAB are mostly advocated recently by researchers and clinicians [5-7]. The technique of decompression by fenestration is a conservative treatment for large cystic jaw masses which is frequently used in recent years [8, 9]. This technique is to create an opening and reduce the pressure in the cyst [10] cavity. By doing so, the cyst will reduce gradually. This technique can help to preserve the appearance and function of the jaws. However, to our best knowledge, only one case report on the treatment for UAB successfully preserved the mandibular second molar of the patient [11]. If the functional occlusion of the teeth involved can be preserved, the life quality of the patients, especially adolescent patients, can be significantly improved. We seek to help the patients to obtain such a possibility. In this report, the technique of decompression by fenestration was applied for the treatment of six adolescents aging from 13 to 15 whose mandibular second molars were imbedded by large UABs. After eight to ten months after the decompression, the cysts reduced and most bone tissues restored. Then surgeries were done to remove the tumors while the mandibular second molars were preserved. During the follow-up period of one to five years, no recurrences were found with a satisfying prognosis.

Material and Methods

Clinical Data

This study was approved by the ethics committee of the Affiliated Hospital of Chifeng College and carried out in accordance with the World Medical Association Declaration of Helsinki. Six adolescent patients including 4 boys and 2 girls aged from 13 to 15 were received at the Affiliated Hospital of Chifeng University from January 2007 to December 2014. Preliminary body checks for the patients indicated that all six patients had facial asymmetry: four with swelling appeared on the left side of the face and two with on the right side of the face. The mouth examination indicated that the rear of the mandibular first molar swelled on the swelling side, and the second molar erupted on the other side. The X-ray (panoramic radiograph) examination indicated a large penetration area from the mandibular ramus to the root tip of the first molar and the second molar was distal to the root tip of the first molar and the second molar was distal to the root tip of the surface that jaw cystic lesions (Figure 1). On the X-ray film (Figure 1), the area of the cyst cavity was larger than 2.5 cm x 10 cm. Four patients had the third molar at the posterior part of the mandibular ramus and the third molar of the other two patients was congenitally absent.

Treatment

Under local anesthesia, the distal surface of the mandibular first molar was cut through to create an opening in the tumor (see Figure 2). The bone tissues on the surface of the tumor were removed to have a 2.0 cm x 1.0 cm cyst wall. A subsequent pathologic examination on the removed tissue was performed. The cystic fluid inside was removed using a suction pump, and the cystic septa was opened to expose the cyst cavity where saline was used to make clean-up. The volume of the cyst cavity was recorded and the cyst was padding with iodoform gauzes. One week after, the iodoform gauzes were removed and an opening retainer made by using the maxillary and mandibular casts was installed to keep the tumor dry and open. The cyst cavity was washed with saline every day with the retainer on the tumor. Subsequent examinations were performed every two weeks afterward. An X-ray (oral panoramic radiograph) examination was taken every two months to measure the volume of the cyst

cavity. This process lasted for eight months (see Figure 3, 4,5).

Eight months later, the size of the tumors of four patients significantly reduced with the restoration of most bone tissues. The tumors and the third molar (if any) were then removed radically while the mandibular second molar was preserved for eruption. The size of the tumors of the other two patients didn't change significantly in eight months. Hence, the tumor removal surgeries were done at the tenth month after the opening retainer installation. The growth of second molars was monitored for six months after the surgery (see Figure 6,7,8).

After the second molar erupted, the second molar position of three patients was in normal dentition. The second molar position of the other three patients was not in normal dentition. Orthodontic treatments were done to tract the mandibular second molar to its normal position. A follow-up of these patients lasted for five years to monitor the disease including the tumor reoccurrence.

Evaluation of the tumor reduction rate

The volume of the cyst cavity was measured based on the normal saline injected into the cyst cavity. Tumor reduction rate = (Cyst cavity volume at the fenestration – Cyst cavity volume at each re-examination)/Cyst volume at the fenestration

Measurement of the tooth growth (height)

The magnifying power was measured by the X-ray film. Magnifying power in X-ray film = Height of the first molar measured on the X-ray film/Actual height of the first molar

The height is measured based on the relative position between the second molar and the distal root of the first molar (see Figure 9).

The second molar is at the horizontal position of the distal root of the first molar on the X-ray film at the fenestration (A).

The second molar is at the horizontal position of the distal root of the first molar on the X-ray film at each re-examination (A_1) .

Distal marginal ridge of the first molar on the X-ray film (B)

Height = $(AB - A_1 B)/Magnifying power in X-ray film$

Results

The HE staining of the cyst wall indicated the existence of ameloblastoma (Figure 10) in the pathological examination of the six patients. Eight months after the decompression by fenestration, the bone mineral density of the mandible inferior border increased and the trabeculae formed along with the cyst reduction by 48.25% (Table 1). Two months after the tumor was removed, the new bone continued to grow. Six months after, the bone restored to normal state, the second molar erupted 14.93 mm. The second molar of three patients was at the buccal side and distal, and the molar returned to the normal dentition after orthodontic treatment (Figure 11). No tumor reoccurrence was observed in the follow-up period of one to five years and the mandibular second molars functioned well.

Discussion

In this article, we report 6 successful surgical cases where patients with UAB achieved full recovery from the disease in the follow-up period of 5 years. These patients received decompression by fenestration surgery followed by the complete removal of the tumor tissue. 8 months after the fenestration, the cyst size decreased 48% while the mandibular second molar grows 6.24 mm. 6 months after the complete removal of the tumor, the cyst disappeared and the patient achieves full recovery with mandibular second molar growing 14.93 mm. Reoccurrence was not found in either of the 6 patients in the 5-year follow-up period. Further, these patients had full recovery on the mandibular second molar function. This report verifies the success of fenestration followed by tumor removal reported by Kazuo et al [11].

Ameloblastoma is classified into, solid/multicystic ameloblastoma, peripheral ameloblastoma, desmoplastic ameloblastoma, and unicystic ameloblastoma according to the World Health Organization[3, 12]. Among them, about 10% of all ameloblastomas are of the unicystic type. Clinically single curettage or segmental resection was usually used as therapeutic options. Recently, decompression by fenestration plus curettage and marsupialization plus curettage has become the two favorable methods for the treatment of UAB [5-7].

Marsupialization is to create a pouch after fenestration and make the cyst cavity expose to the air in order to promote the natural eruption of the tooth within the cyst. This method is usually used when teeth are involved and wrapped in the cyst [13-15]. Hyomoto et al. [16] performed marsupialization for 58 patients having cysts with teeth involved, and the teeth of 42 patients had good eruption. Kazuo Sano [11] reported one case of mandible UA treated by marsupialization followed by enucleation without removal of an involved second molar. In this case, the patient had an eruption of the mandibular second molar one year after the surgery. In the follow-up period of another 51 months, the patient obtained occlusion without tumor reoccurrence. The technique of decompression by fenestration is applied for the treatment of cysts within large jaws [8, 9]. The method is to eliminate the destruction of the bones by creating and maintaining an opening on the surface of the large cystic lesion within the jaw where the pressure within the cyst cavity reduces to a normal level. The bone tissue would be benefit from the opening to achieve recovery [17]. The difference between the fenestration and the marsupialization is that a pouch is not created but an opening retainer is put to keep the cyst cavity communicating with the air. A negative pressure suction pump can also be installed to accelerate the reduction of cysts. In this case, we employed the technique of decompression by fenestration and performed tumor enucleation surgery while preserving the mandibular second molar. We successfully obtained the same result as that of Kazuo Sano [11]: patients' face was kept well and the function of jaw and toot was also preserved. During the follow-up period of five years, there was no tumor reoccurrence and the mandibular second molar functioned well.

However, not all affected teeth within UABs can be preserved in conservative treatments. Hyomoto et al. [16] pointed out that the merits of marsupialization were to reduce the cyst cavity and preserve the teeth within the cyst. However, if the tooth apical foramen within the cyst was formed, the tooth could not erupt. In our practice, to preserve the impacted mandibular second molar within UAB, the following four conditions are needed: first, fenestration and decompression or marsupialization has to be performed to reduce the cyst cavity; second, the tumor must be removed radically; third, the apical foramen of the affected second molar is formed; fourth, the second molar is not occluded by adjacent teeth and there is enough space for eruption.

UABs are histologically classified into 3 subtypes: unilocular cystic luminal (subtype I), intraluminal with a solid growth inside lumen of the cystic lesion (subtype II); and intraluminal growth with mural invasion within adjacent tissues (mural AB, or subtype III). Lee et al. [18] employed single enucleation to treat 22 patients with UAB including five mural AB cases. During the follow-up period of four years, the tumor reoccurred in two patients, and these two were both mural AB. They suggested following up for a long time after treatment. In this study, among the six cases, four patients had UAB of unilocular cystic luminal and two patients had UAB of intraluminal with a solid growth inside lumen of the cystic lesion. A study suggested that the average interval to recurrence of UAB was approximately 7 years [19]. Although the follow-up period in this study is five years without reoccurrence, a longer time of follow-up is needed.

Conclusion

UAB is likely to occur in adolescents, who attach importance to their appearance. It is our suggestion to apply the technique of decompression by fenestration plus tumor removal while preserving the mandibular second molar. This method preserves the functions of jaw and tooth involved and has the minimum effect on patients' appearance. Although the number of cases in this study is limited and the reliability of the results is needed to be verified, this method can remove the tumor and reduce the influence on adolescent patients' psychology burden at the same time. It can be a favorable treatment for the disease with strict and long-time follow-up.

Figure legends



Figure 1 X-ray film of ameloblastoma before surgery.



Figure 2 Tumor fenestration surgery.



Figure 3 X-ray films of two to eight months after surgery.



Figure 4 X-ray films of four months after surgery.



Figure 5 X-ray films of six months after surgery.



Figure 6 X-ray films of two months after tumor removal.



Figure 7 X-ray films of four months after tumor removal.



Figure 8 X-ray films of six months after tumor removal.



Figure 9 X-ray film after orthodontic treatment.



Figure 10 Relative position between the second molar and the distal root of the first molar.



Figure 11 HE staining result of UAB (X200). The manification bar represents 50µm.

Acknowledgements

This study was supported by the grants from the Inner Mongolia Natural Science Foundation of China (Grand No. 2016MS0808).

Running title: Fenestration for unicystic ameloblastoma treatment

References

- [1] Li, W.C., et al., *Expression and significance of survivin and COX- 2 in ameloblastoma*. Journal Practical Stomatology, 2014. **30**(1): p. 126-128.
- [2] Neville, B.W., et al., Oral and Maxillofacial Pathology. 2008, Saunders. p. 708.
- [3] Gunawardhana, K.S., P.R. Jayasooriya, and W.M. Tilakaratne, Diagnostic dilemma of

unicystic ameloblastoma: novel parameters to differentiate unicystic ameloblastoma from common odontogenic cysts. J Investig Clin Dent, 2014. **5**(3): p. 220-5.

- [4] Seintou, A., C.P. Martinelli-Klay, and T. Lombardi, Unicystic ameloblastoma in children: systematic review of clinicopathological features and treatment outcomes. Int J Oral Maxillofac Surg, 2014. 43(4): p. 405-12.
- [5] Tomita, Y., et al., Orthodontic occlusal reconstruction after conservative treatment of unicystic ameloblastoma in an adolescent patient: 10-year follow-up. Am J Orthod Dentofacial Orthop, 2013. 144(3): p. 466-70.
- [6] Bisinelli, J.C., et al., *Conservative treatment of unicystic ameloblastoma*. Am J Orthod Dentofacial Orthop, 2010. 137(3): p. 396-400.
- [7] Dolanmaz, D., et al., *Marsupialization of unicystic ameloblastoma: a conservative approach for aggressive odontogenic tumors.* Indian J Dent Res, 2011. **22**(5): p. 709-12.
- [8] Enislidis, G., et al., Conservative treatment of large cystic lesions of the mandible: a prospective study of the effect of decompression. Br J Oral Maxillofac Surg, 2004. 42(6):
 p. 546-50.
- [9] Hu, Y.J., S.Y. Li, and C.P. Zhang. The clinical research of decompression treatment of large mandibular cystic lesions. in Proceedings of the Symposium on the New Progress in the Diagnosis, Treatment and Reconstruction of Oral Cancer in the Cross-Strait. 2008.
- [10] Xavier, S.P., et al., Conservative approach: using decompression procedure for management of a large unicystic ameloblastoma of the mandible. J Craniofac Surg, 2014.
 25(3): p. 1012-4.
- [11] Sano, K., et al., Spontaneous eruption of involved second molar in unicystic ameloblastoma of the mandible after marsupialization followed by enucleation: a case report. J Oral Maxillofac Surg, 2013. 71(1): p. 66-71.
- [12] Barnes, L., et al., World Health Organization Classification of Tumours: Pathology and Genetics of Head and Neck Tumours. Vol. 9. 2005: IARC. 299-300.
- [13] Ertas, U. and M.S. Yavuz, Interesting eruption of 4 teeth associated with a large dentigerous cyst in mandible by only marsupialization. J Oral Maxillofac Surg, 2003.
 61(6): p. 728-30.
- [14] Fujii, R., et al., Panoramic findings for predicting eruption of mandibular premolars

associated with dentigerous cyst after marsupialization. J Oral Maxillofac Surg, 2008. **66**(2): p. 272-6.

- [15] Yahara, Y., et al., Eruption prediction of mandibular premolars associated with dentigerous cysts. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 2009. 108(1): p. 28-31.
- [16] Hyomoto, M., et al., Clinical conditions for eruption of maxillary canines and mandibular premolars associated with dentigerous cysts. Am J Orthod Dentofacial Orthop, 2003. 124(5): p. 515-20.
- [17] Zhou, C.X., Z.M. Huang, and B.L. Liang, AProach to Roentgenologic Diagnosis of Ameloblastoma of the Jaw:Analysis of 64 Cases. Journal of Diagnostic Imaging & Interventional Radiology, 1998(2): p. 83-86.
- [18] Lee, P.K., N. Samman, and I.O. Ng, Unicystic ameloblastoma--use of Carnoy's solution after enucleation. Int J Oral Maxillofac Surg, 2004. 33(3): p. 263-7.
- [19] Li, T.J., et al., Unicystic ameloblastoma: a clinicopathologic study of 33 Chinese patients.Am J Surg Pathol, 2000. 24(10): p. 1385-92.